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Agressive osteoblastoma in a seven-year-old girl's mandible: Treatment and six-year monitoring



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ABSTRACT

INTRODUCTION: Osteoblastoma is a rare benign bone tumor with locally aggressive behavior in some cases; however, with rare malignant transformations. A case of osteoblastoma in a seven-year-old patient's mandible is presented.

PRESENTATION OF CASE: After the diagnosis, the patient underwent two stages of treatment. In the first intervention, an intralesional curettage was performed as well as a marsupialization of the lesion, in order to reduce its volume and allow a second and more conservative surgical period. In the second surgery, after reducing the lesion, enucleation and rigid internal fixation of the mandible were performed.

DISCUSSION: The treatment has proven to be satisfactory, without recurrence of the injury for two years, when the patient suffered a car accident that caused the synthetic material to fracture. Conclusion: The internal fixation was surgically replaced and the patient is being monitored after six years of the initial intervention, without presenting further changes.

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1. Introduction

Osteoblastoma is an uncommon benign bone tumor and accounts for less than one percent of all bone tumors [1]. The most frequently affected bones are the backbone, sacrum, calvaria, the longer bones, and small bones of the hands and feet [2]. Approximately 15% of the cases occur in the maxillofacial area, with a higher frequency of occurrence in the mandible [3].

In radiographs, the lesion may appear radiolucent, well or poorly defined, and containing variable amounts of mineralization [4].

Pain and swelling of the affected area are typical features presented by osteoblastoma in the facial bones. However, the damage can be discovered on routine clinical examination, while it shows no signal or causes any symptoms [5].

In both clinical and histological terms, a distinctive diagnosis for osteoblastomas varies between benign and malignant tumors, such as cementoblastoma, osteoid osteoma, fibrous dysplasia, ossifying fibroma, focal cemento-osseous dysplasia to low-grade osteosarcoma [6].

The treatment for osteoblastoma is defined by a complete excision of the lesion or curettage [6]. There are reports of regression after biopsy or incomplete removal, suggesting that the osteoblastoma may be a reactive process [7].

This study describes a case of osteoblastoma in a seven-year-old patient's mandible, her six-year treatment and monitoring.

2. Case report

Female patient, leucoderma, seven years old, was referred from the Department of Pediatric Oncology from Hospital de Câncer de Mato Grosso to the Department of Dentistry of the same institution for the purposes of assessment and management. The patient was accompanied by her father and reported as the main complaint an abnormal swelling in her face, with six months of development and pain in the area. No other morbid medical history of clinical relevance reported.

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Fig. 1. Initial image of the patient in front (A) and inferior/superior (B) view. The patient showed facial asymmetry with volumetric increase in the submandibular and left parotid-masseteric region, extending to the ipsilateral neck region, hardened, with resilient and crackling areas.

When the headgear physical examination was performed, an asymmetrical face with volumetric increase in the submandibular and left parotid-masseteric region extending to the ipsilateral neck region, hardened, with resilient and crackling areas, painful on palpation and with intact integument were detected (Fig. 1).

The intraoral physical examination revealed healthy, colored mucous membranes, and obliteration of the lower gingival sulcus on the left. Patient presented mixed dentition, with some cavitated teeth. However, in the lower left quadrant, teeth were intact.

The requested additional tests were panoramic radiography (Fig. 2), chest radiography (AP and lateral), computed tomography of the face (Fig. 3), ultrasound of cervical and submandibular areas and routine preoperative examinations (blood count, coagulogram, fasting glycaemia, creatinine, urea, bilirubin, GOT and GPT). The laboratory examinations results were within the normal standards and Class 1 Grade I surgical risk.

Lymphoma, ameloblastoma, keratocystic odontogenic tumor and aneurysmal bone cyst have been proposed as differential diagnoses.

An incisional biopsy was performed with headgear access under local anesthesia with the use of intravenous sedation in a surgical center. The histological analysis revealed a richly vascularized lesion, with many tissue trabeculae with gaps filled by osteocytes, osteoid matrix, osteoblasts and giant multinucleated cells leading to the diagnosis of osteoblastoma (Fig. 4).

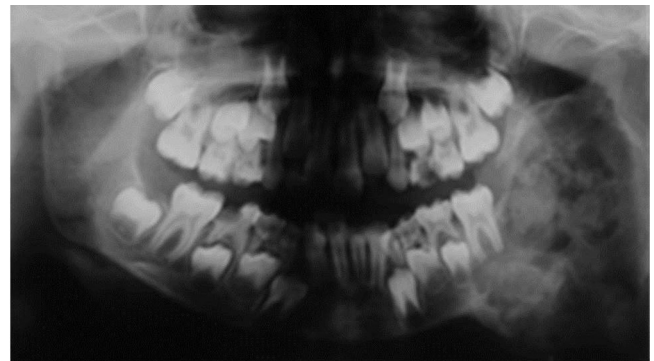


Fig. 2. Initial panoramic radiograph showing patient in mixed dentition and presence of destructive lesion with poorly defined margins and a predominantly mixed radiographic pattern in the left mandibular body, angle and ramus.

Considering the age of the patient, a decision was made: To perform the marsupialization of the lesion with intralesional curettage, followed by a cyst removal technique with furacin gauze, in order to reduce its volume and allow a second and more conservative surgical procedure (Fig. 5). During surgery, a pathological fracture in the left mandibular angle occurred, due to the large extent of the lesion and the remaining bone fragility, however, the rigid inter-

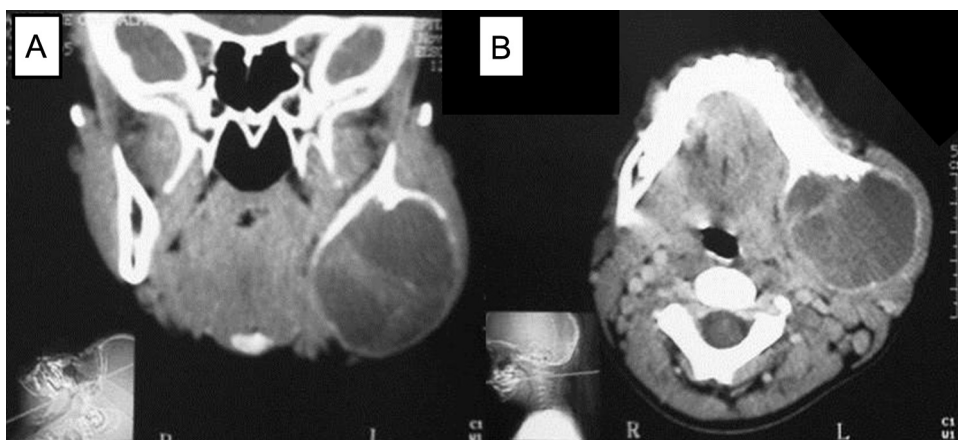


Fig. 3. Computed tomography in coronal (A) and axial (B) sections showing an hypodense area of approximately 8 mm in its largest diameter in the mandibular angle and left mandibular body areas, well-defined margins, cortical bone expansion and solid-cystic component.

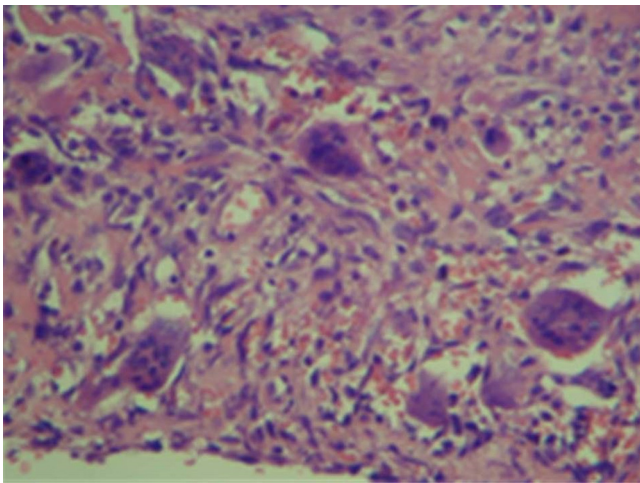


Fig. 4. Multinucleated giant cell groups near small hemorrhage focal points, accompanied by mononuclear cells. Formation of reactive bone inside fusiform cells stroma.

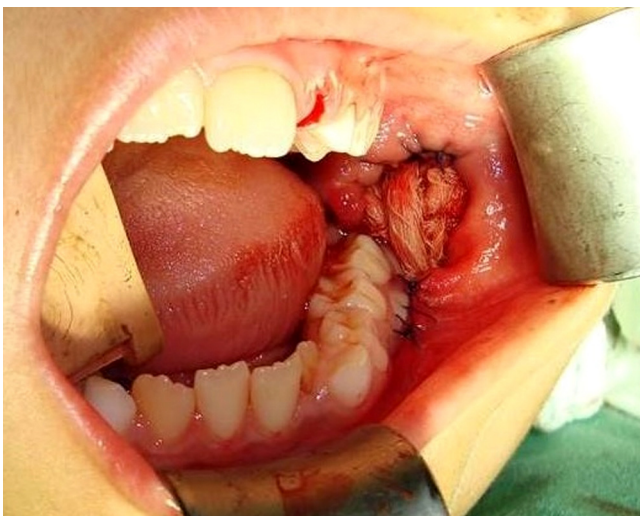


Fig. 5. Clinical view of the lesion with furacin gauze after marsupialization.

nal fixation was not performed, taking into account the occlusal stability and need for another surgical procedure for the complete removal of the lesion.

After three months, the lesion had decreased 24 mm, spanning to 56 mm in its greatest dimension (Fig. 6). Thus, it was enucleated through submandibular access combined with intraoral access, curettage and cryotherapy. In order to promote the fixation of the mandible, titanium plate and screws were used. No nerve damage was observed after surgery.

The patient progressed, having a good response in the postoperative, returning in 7, 15, 30, 60, 90 and 120 days.

After two years, the patient was involved in a car accident, fracturing the synthesis material (Fig. 7), and underwent a new surgery for replacement of the plate and of the 2.4 mm titanium screws.

The patient progressed, having a good response in the postoperative, returning in 7, 15, 30, 60, 90 and 120 days.

Currently six years have passed since the curettage of osteoblastoma in the patient's mandible and four years have passed since the surgery for exchanging the plate. The patient is in a good general state with no recurrence of the lesion (Fig. 8).

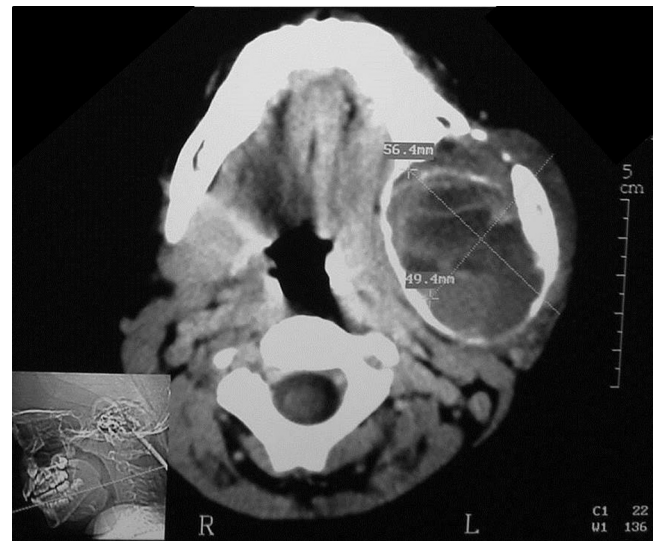


Fig. 6. Computed tomography in axial section showing a decreased lytic area in the order of 30% after marsupialization.

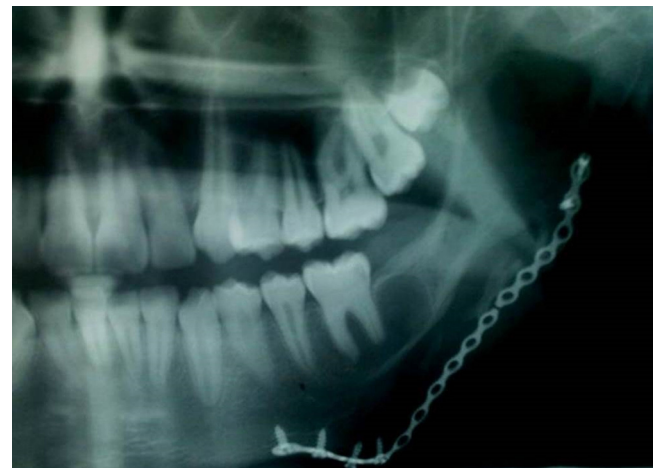


Fig. 7. Panoramic radiograph showing of the fracture of the synthesis material.

3. Discussion

Jaffe and Mayer described the osteoblastoma in 1932 under the name "Osteoblastic and osteoid tissue-forming tumor". Dahlin and Johnson in 1954 called it "giant osteoid osteoma". In 1956, Jaffe and Lichtenstein detailed the clinical and histopathological characteristics of the osteoblastoma. The first case of osteoblastoma in the mandible was described by Barelo and Sedano in 1967 [3].

This rare tumor is histologically benign, osteoid-forming vascular neoplasm [8] and accounts for less than 1% of all bone lesions, and 15% of these tumors affect the head region [9]. It occurs twice as often in men, and usually in their mid-thirties. Vertebrae and long bones stand out as its preferred sites [10]. In the presented case the lesion occurred in the mandible, in a female seven-year-old patient, an unusual fact.

The volume increase may precede the development of pain, as observed in this case, which may or may not be relieved by non-steroidal anti-inflammatory drugs [11]. Most of the tumors start growing in the bone marrow [5], as in this case.

In the radiography, the osteoblastoma has an oval or round image which is radiolucent, radiopaque or mixed, however, without fusing with the cementum of the adjacent teeth, sometimes surrounded by a sclerotic bone margin and in approximately



Fig. 8. Patient in the six-year postoperative period in front view (A) and profile (B) presenting a symmetrical face and scar in submandibular region.

one third of cases resembles a malignant pathology such as the osteosarcoma [3,10]. In this case a mixed pattern, multilocular image, bulging of the bone cortical bone, with a moth-eaten aspect, was observed.

In histological aspects, the osteoid osteoma and the osteoblastoma are similar [6]. They consist of a mineralized matrix (osteoid tissue islands with varying degrees of calcification) with numerous hyperchromatic osteoblasts and occasionally giant cells similar to osteoclasts in a highly vascularized fibrous stroma [12]. By virtue of being histologically similar, the diagnosis between osteoblastoma and osteoid osteoma is based on the diameter of the lesion, since the latter has a limited potential for growth (15–20 mm) when compared to the first [13].

Conventional osteoblastomas have their growth potential limited, and usually do not exceed 40 mm in diameter [14]. They grow slowly over many years and have a well-defined sclerotic margin, are fairly well vascularized with a mild inflammatory response [15]. In this case, the lesion had a diameter of 80 mm, which is characteristic of an aggressive osteoblastoma, which exhibits locally aggressive behavior with a propensity to recur [15].

Differentiating between the conventional and aggressive osteoblastomas is important to determine the appropriate treatment and prevent recurrence. The recommended treatment is surgical resection with safety margins and reconstruction [5,15,16], although computed tomography-guided percutaneous cryoablation [1] and polymethylmethacrylate filling of the resulting gap [17] have also been proposed. The proposed treatment established in this case was in two stages, for the injury manifested aggressively destroying the cortical bones that supported the mandibular body in the framework region, angle and left ascending branch. Also, a more aggressive treatment could result in significant loss of the surrounding bone and the need for primary reconstruction in order to maintain an aesthetic facial contour, and the masticatory functions. Another factor considered was the fact that the lesion responds to curettage, which is a conservative treatment [4,12] and also because the patient is a child. The treatment was satisfactory, presenting areas of new bone formation with three months of development analyzed, configuring an area with resistance and sustainability of the lines of force pertaining to the remaining bone decreasing the risk of a new pathologic fracture, and preventing reconstruction with bone graft in the curetted region.

Recurrence of osteoblastomas was recorded in 13.6% of cases [4]. After six years of control, which involved the replacement of the reconstruction material due to the fracture in a car accident, the patient is in good state without recurrence and new bone formation throughout the extension of the mandible.

Conflict of interest

The authors do not have any financial and personal relationships with other people or organisations that could inappropriately influence (bias) this work.

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Ethical approval

Does not apply.

Consent

Written informed consent was obtained from the parent of the patient for publication of this case report and the accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Paulo Henrique Souza Castro participated in guiding the design and clinical conduct of the case presented and critically reviewed the manuscript.

Danielle Lima Molinari participated in the clinical management of the case and the writing of the manuscript.

Hiran Queiroz Stateri participated in the clinical management of the case and the writing of the manuscript.

Alvaro Henrique Borges guided the writing of the manuscript, translated the text into English, selected the journal and formatted the work in publishing standards.

Luiz Evaristo Ricci Volpato guided the whole process of writing and critical discussion of the article.

Guarantor

Luiz Evaristo Ricci Volpato and Paulo Henrique de Souza Castro.

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